

A preliminary examination of self-concept in older adolescents and young adults with Gilles de la Tourette syndrome

Silvestri, Paola R.; Chiarotti, Flavia; Baglioni, Valentina; Neri, Valeria; Cardona, Francesco; Cavanna, Andrea E.

DOI:

[10.1016/j.ejpn.2016.12.006](https://doi.org/10.1016/j.ejpn.2016.12.006)

License:

Creative Commons: Attribution-NonCommercial-NoDerivs (CC BY-NC-ND)

Document Version

Peer reviewed version

Citation for published version (Harvard):

Silvestri, PR, Chiarotti, F, Baglioni, V, Neri, V, Cardona, F & Cavanna, AE 2016, 'A preliminary examination of self-concept in older adolescents and young adults with Gilles de la Tourette syndrome', *European Journal of Paediatric Neurology*. <https://doi.org/10.1016/j.ejpn.2016.12.006>

[Link to publication on Research at Birmingham portal](#)

General rights

Unless a licence is specified above, all rights (including copyright and moral rights) in this document are retained by the authors and/or the copyright holders. The express permission of the copyright holder must be obtained for any use of this material other than for purposes permitted by law.

- Users may freely distribute the URL that is used to identify this publication.
- Users may download and/or print one copy of the publication from the University of Birmingham research portal for the purpose of private study or non-commercial research.
- User may use extracts from the document in line with the concept of 'fair dealing' under the Copyright, Designs and Patents Act 1988 (?)
- Users may not further distribute the material nor use it for the purposes of commercial gain.

Where a licence is displayed above, please note the terms and conditions of the licence govern your use of this document.

When citing, please reference the published version.

Take down policy

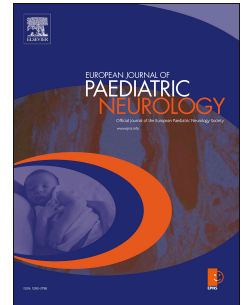
While the University of Birmingham exercises care and attention in making items available there are rare occasions when an item has been uploaded in error or has been deemed to be commercially or otherwise sensitive.

If you believe that this is the case for this document, please contact UBIRA@lists.bham.ac.uk providing details and we will remove access to the work immediately and investigate.

Accepted Manuscript

A preliminary examination of self-concept in older adolescents and young adults with Gilles de la Tourette syndrome

Paola R. Silvestri, Flavia Chiarotti, Valentina Baglioni, Valeria Neri, Francesco Cardona, Andrea E. Cavanna, MD PhD FRCP, Prof.



PII: S1090-3798(16)30283-5

DOI: [10.1016/j.ejpn.2016.12.006](https://doi.org/10.1016/j.ejpn.2016.12.006)

Reference: YEJPN 2163

To appear in: *European Journal of Paediatric Neurology*

Received Date: 22 June 2016

Revised Date: 5 December 2016

Accepted Date: 11 December 2016

Please cite this article as: Silvestri PR, Chiarotti F, Baglioni V, Neri V, Cardona F, Cavanna AE, A preliminary examination of self-concept in older adolescents and young adults with Gilles de la Tourette syndrome, *European Journal of Paediatric Neurology* (2017), doi: 10.1016/j.ejpn.2016.12.006.

This is a PDF file of an unedited manuscript that has been accepted for publication. As a service to our customers we are providing this early version of the manuscript. The manuscript will undergo copyediting, typesetting, and review of the resulting proof before it is published in its final form. Please note that during the production process errors may be discovered which could affect the content, and all legal disclaimers that apply to the journal pertain.

**A preliminary examination of self-concept in older adolescents and young adults with
Gilles de la Tourette syndrome**

Paola R. Silvestri¹, Flavia Chiarotti², Valentina Baglioni¹, Valeria Neri¹, Francesco Cardona¹,
Andrea E. Cavanna^{3,4,5*}

¹Department of Pediatrics and Child Neuropsychiatry, Sapienza University, Rome, Italy

²Department of Cell Biology and Neuroscience, National Institute of Health, Rome, Italy

³Department of Neuropsychiatry, BSMHFT and University of Birmingham, Birmingham, United Kingdom

⁴Sobell Department of Motor Neuroscience and Movement Disorders, Institute of Neurology and University College London, London, United Kingdom

⁵School of Life and Health Sciences, Aston Brain Centre, Aston University, Birmingham, United Kingdom

* Corresponding author

Prof. Andrea E. Cavanna, MD PhD FRCP

Department of Neuropsychiatry

The Barberry National Centre for Mental Health

25 Vincent Drive

Birmingham B15 2FG

E-mail: a.cavanna@ion.ucl.ac.uk

Abstract

Gilles de la Tourette syndrome (GTS) is a childhood-onset neuropsychiatric disorder characterised by multiple tics and often associated with behavioural problems. Although there is evidence of significantly reduced self-esteem in children and adolescents with GTS, little is known about perceived self-concept and its clinical determinants at the transition age between adolescence and adulthood. We therefore set out to investigate self-concept in a clinical sample of young patients with GTS at this crucial age for personal development. In addition to standard demographic and clinical data, we collected self-ratings using a standardised battery of psychometric instruments, as well as the Multidimensional Self Concept Scale, a comprehensive questionnaire developed to assess self-concept in subjects aged 9 to 19 years, tapping into the social, competence, affect, academic, family, and physical domains. We found that patients diagnosed with at least one co-morbid psychiatric disorder (“GTS-plus” phenotype) reported significantly lower self-concept than patients with “pure GTS”, whereas tic-related variables had no impact on self-concept. Anxiety symptoms were the main determinants of self-concept, especially trait anxiety with regard to social and affective domains. Affective symptoms could also have a negative impact on the physical, affective, competence, and social domains of self-concept. Routine screening for these co-morbid conditions should be recommended in all patients with GTS seen at transition clinics from paediatric to adult care, in order to implement effective treatment interventions whenever possible.

Key words: anxiety; depression; Gilles de la Tourette syndrome; psychiatric co-morbidities; self-concept; tics.

Introduction

Gilles de la Tourette syndrome (GTS) is a childhood-onset neuropsychiatric disorder characterised by multiple motor tics plus at least one phonic tic lasting longer than one year^(1,2). The average age at onset of GTS is around 7 years. The prevalence of GTS in children and adolescents is up to 1%, with a male:female ratio of 3-4:1, and a significant reduction of tic severity after adolescence occurs in about one third of cases^(3,4). Co-morbid psychiatric disorders are reported by the vast majority (90%) of patients with GTS, especially attention-deficit hyperactivity disorder (ADHD) and obsessive-compulsive disorder (OCD)⁽²⁾. GTS is also associated with anxiety and affective disorders, rage attacks, impulsivity and self-injurious behaviours⁽⁵⁻⁷⁾.

Both tic severity and the presence of psychiatric co-morbidities have been shown to affect patients' health-related quality of life (HR-QoL)^(8,9). Tics can impair everyday activities (including job and school attendance) and social relationships⁽¹⁰⁾. School functioning is often compromised, especially in young patients with co-morbid ADHD⁽¹¹⁾. Although co-morbid ADHD is known as the main cause of social and behavioural difficulties in this patient population⁽¹²⁾, affective symptoms have consistently been shown to be a major determinant of patients' wellbeing, as a considerable proportion of patients with GTS receive a formal diagnosis of affective disorder, irrespective of being diagnosed with ADHD, throughout adolescence⁽¹³⁾ and adulthood⁽¹⁴⁾. In particular, there is evidence of significantly reduced self-esteem in children and adolescents with GTS. A study by Khalifa et al.⁽¹⁵⁾ showed that children with GTS have lower self-perception compared to unaffected children. Children with GTS and co-morbid ADHD reported the most significant impairment in the physical appearance and social interactions domains. Moreover, poor self-perception in the physical appearance domain was found to be associated with earlier age at tic onset. Hesapçioğlu et al.⁽¹⁶⁾ found that female patients younger than 12 years reported lower self-esteem because of their tic disorder. Low self-esteem was found to be associated with impairment in all HR-QoL areas, with the exception of the academic domain. Although the concepts of self-esteem and self-concept have often been used interchangeably, Bracken specifically defined individuals' self-concepts as "learned evaluations of themselves that are based upon their past successes and failures, reinforcement histories, and the ways others react to them and interact with them"⁽¹⁷⁾. Self-esteem and self-concept are components of self-perception which affect the broader concept of HR-QoL. A study by Hanks et al.⁽¹⁸⁾ showed that children and adolescents with a diagnosis of chronic tic disorder have reduced levels of self-concept, and identified a negative correlation between self-concept and the severity of both tics and co-morbid psychiatric disorders. An association was observed between

patient's self-concept and overall HR-QoL, and it was noted that younger children are less likely to exhibit depressive symptoms and tend to report higher self-concepts than adolescents.

To date, few data are available about the impact of GTS at the transition between adolescence and adulthood, a crucial age for the development of self-concept. This is a delicate age for patients with tic disorders, as the persistence of symptoms into young adulthood suggests that patients might have to face the possibility that their tic disorder persists throughout life as a potentially stigmatising condition. Self-concept is particularly important amongst older adolescents with tics, who represent a group of young people with GTS at a crucial point in their lives both in terms of adjustment and maturation and also in relation to their being at a watershed for the progression or decline of symptoms. As tics will remit for many children by late adolescence, those who have tics in late adolescence will likely have them for the remainder of their lives. As self-concept is important for long-term functional outcomes (e.g. job, relationships, social factors), the importance of understanding factors that contribute to self-concept during this key developmental period cannot be underestimated. We set out to investigate self-concept and its determinants in a clinical sample of adolescents and young adults with GTS. This was the first study to focus on late adolescence: we hypothesised that patients with GTS in this age group experience problems with self-concept related to co-morbid conditions ⁽¹⁹⁾ and assessed their clinical correlates with a comprehensive battery of psychometric instruments.

Methods

Participants

Twenty-two patients (5 girls) with a diagnosis of GTS and a mean age of 18 years (age range 15-19 years; standard deviation (SD) 1 year), were recruited from the Child and Adolescent Neuropsychiatry Outpatient Unit of the Sapienza University in Rome between October 2013 and February 2015. The main inclusion criterion was the presence of a validated diagnosis of GTS according to current Diagnostic and Statistical Manual for Mental Disorders (DSM) criteria ⁽¹⁾. Exclusion criteria consisted in the diagnosis of intellectual disability, schizophrenia or other psychotic disorders. Suitable participants were recruited from consecutive attenders to our specialist clinic and included a mix of newly referred patients and follow-up patients. Each participant and his/her parents (for patients younger than 18 years) provided written informed consent before enrollment.

All participants were of white Caucasian ethnic origin with Italian as first language. All but 5 participants (of which 3 with vocational training diploma) were still in education and living with their parents. No patient had educational support needs or physical disorders. Co-morbid diagnoses were established through clinical interview incorporating previous diagnostic records. Eight out of the 22 participants had no co-morbid psychiatric disorder ("pure" GTS), 8 were diagnosed with OCD, 4 with ADHD, and 2 with both OCD and ADHD. The mean age at tic onset was 8 years (SD 3 years). Five participants had at least one first-degree relative with a tic disorder. Eighteen participants were on pharmacotherapy (8 with atypical antipsychotics, 1 with typical antipsychotics, 2 with antidepressants, 7 with other medications). The duration of pharmacotherapy ranged from 1 month to 8 years and there were no previous psychological and pharmacological therapeutic interventions other than the ones noted on assessment. Mean duration of education was 11 years (SD 1 year).

Procedure

Each participant was systematically assessed within a single session. Specialists in child and adolescence psychiatry with extensive experience in tic disorders collected demographic and clinical information (ethnicity, first language, gender, age, age at tic onset, family history of tics, educational attainment, employment), reviewed inclusion and exclusion criteria, and assessed the severity of tics and obsessive-compulsive symptoms using the following two validated rating scales:

- *Yale Global Tic Severity Scale (YGTSS)* ⁽²⁰⁾

The YGTSS is a clinician-rated instrument that assesses the number, frequency, intensity, complexity and interference of motor and phonic tics. YGTSS total tic severity scores range from 0 (no tics) to 50 (extremely severe tics).

- *Yale-Brown Obsessive-Compulsive Scale (Y-BOCS)* ⁽²¹⁾

The Y-BOCS is a clinician-rated instrument that assesses obsessions and compulsions. Each item receives a score from 0 to 5 and total scores range from 0 (no obsessive-compulsive symptoms) to 40 (extremely disabling obsessive-compulsive symptoms).

Each participant was subsequently asked to complete a standardised battery of psychometric instruments validated in the Italian language to assess premonitory urges, self-concept, and co-morbid psychiatric symptoms. The choice of psychometric instruments was guided by the available evidence on valid measures in this patient population ^(22,23).

- *Premonitory Urge for Tics Scale (PUTS)* ⁽²⁴⁾

The PUTS is a 10-items self-report scale that assesses premonitory urges to tic. Items from 1 to 9 receive a score ranging from 1 (never) to 4 (always). Higher scores indicate the presence of more severe urges.

- *Multidimensional Self Concept Scale (MSCS)* ⁽¹⁷⁾

The MSCS is a 150-item, self-report questionnaire developed to assess self-concept in subjects aged 9 to 19 years based on Bracken's multidimensional self-concept model. The MSCS covers 6 content-dependent self-concept areas, tapping into the social, competence, affect, academic, family, and physical domains. Each item is rated on a 4-point scale and higher scores indicate better self-concept. In addition to general populations, the MSCS has been used in previous studies to evaluate self-concept in patients with chronic neuropsychiatric conditions ^(25,26).

- *Multidimensional Anxiety Scale for Children (MASC)* ⁽²⁷⁾

The MASC assesses anxiety through a 39-item, self-report measure for use in subjects aged 8 to 19 years. Items are rated on a 4-point Likert-type scale, and higher scores indicate more severe symptoms.

- *State-Trait Anxiety Inventory-form Y (STAI-Y)* ⁽²⁸⁾

The STAI is a 40-item, self-report questionnaire validated to assess state anxiety (STAI-Y1) and trait anxiety (STAI-Y2) in subjects older than 12 years. Items are rated on a 4-point Likert-type scale, and higher scores indicate more severe symptoms.

- *Beck Depression Inventory-II (BDI-II)* ⁽²⁹⁾

The BDI-II is a 21-item, self-report questionnaire validated to assess symptoms of depression in subjects older than 13 years. Items are rated on a 4-point Likert-type scale, and higher scores

indicate more severe symptoms. Separated scores are provided for cognitive-affective and somatic-vegetative constructs.

Statistical analysis

Quantitative data are presented as mean values (\pm SD), while categorical data are presented as absolute and percent frequencies. For the analysis, participants were divided in subgroups based on the following grouping variables: (a) gender and other dichotomous variables (e.g. male *vs* female; presence *vs* absence of family history of tic disorders); (b) tic severity (YGTSS score <10 *vs* ≥ 10 , where 10 is the cut-off between minimal and mild tic severity); (c) premonitory urge severity (PUTS score <25 *vs* ≥ 25 , where 25 is the cut-off between medium and high intensity of premonitory urges for tics); (d) anxiety symptoms (MASC score <50 *vs* ≥ 50), anxiety state (STAI-Y1 $<75^{\text{th}}$ centile *vs* $\geq 75^{\text{th}}$ centile) and trait (STAI-Y2 $<75^{\text{th}}$ centile *vs* $\geq 75^{\text{th}}$ centile); (e) affective symptoms (BDI-II $<90^{\text{th}}$ centile *vs* $\geq 90^{\text{th}}$ centile); (f) phenotype (“pure” GTS *vs* “GTS-plus”, i.e. GTS with co-morbid psychiatric disorders). Differences in MSCS scores among subgroups were assessed using the Mann-Whitney U test and Kruskal-Wallis analysis of variance (for two or more than two independent groups, respectively). Finally, multiple linear regression analysis was performed to determine the variables significantly and independently affecting MSCS total scores. For any dependent variable different models were fitted to data: the adjusted R-squared and Akaike Information Criterion (AIC) were computed as measures of model goodness-of-fit. The variance inflation factor (VIF) was computed to assess the presence of relevant multicollinearity among predictors within a model, with VIF values equal to or greater than 5 suggesting multicollinearity. We presented the results of the multiple regression analysis for the model showing the best compromise between AIC and VIF values (for both, the lower the value, the better the model). Cohen’s f^2 values were also calculated to compare the effect sizes of the different predictors within each model: $f^2 \geq 0.02$, $f^2 \geq 0.15$, and $f^2 \geq 0.35$ represent small, medium, and large effect sizes, respectively. We reported statistically significant regression coefficients, expressed as variation in the dependent variable corresponding to a one-unit increase in the predictor score, for any predictor alongside the corresponding Cohen’s f^2 . Statistical analyses were performed using the STATA Statistical Software, Version 8.1.

Results

In our sample, MSCS total and subscale scores were lower in patients with GTS and co-morbid psychiatric disorders compared to patients with “pure” GTS: differences were particularly marked in patients who reported more severe anxiety symptoms (as rated by both STAI and MASC scores) (**Table 1**).

[PLEASE INSERT TABLE 1 HERE]

Patients with at least one co-morbid psychiatric diagnosis reported significantly lower self-concept, with more pronounced effects on the competence, academic, affect, and social MSCS domains. Specifically, the presence of more severe anxiety and affective symptoms was consistently associated with significantly lower MSCS social, affect, competence, and physical scores; in addition, the presence of more severe state/trait anxiety symptoms were associated with significantly lower MSCS family and academic scores. No significant differences in MSCS scores were observed between male and female patients, or between patients with lower or higher tic/premonitory urge severity. Likewise, neither socio-demographic variables nor treatment interventions were associated with decreased self-concept in our clinical sample.

R-squared values resulting from the multiple linear regression analysis on MSCS scores were consistently high, ranging from 0.59 to 0.84, with the exception of MSCS family scores (R-squared 0.34) (**Table 2**).

[PLEASE INSERT TABLE 2 HERE]

Trait anxiety (STAI-Y2) and, to a lesser extent, depression (BDI-II) centile scores negatively affected specific self-concept (MSCS) subscale scores. Specifically, a one-unit increase in the STAI-Y2 centile score determined a significant estimated mean decrease of 0.29 in the MSCS total score ($f^2=0.39$), 0.37 in the MSCS social score ($f^2=0.19$), 0.27 in the MSCS competence score ($f^2=0.28$), 0.22 in the MSCS affect score ($f^2=0.15$), and 0.23 in the MSCS family score ($f^2=0.21$). According to Cohen's f^2 values, the effect size was large for the MSCS total score, while it was medium for all subscale scores. A one-unit increase in the BDI-II centile score determined a significant estimated mean decrease of 0.24 in the MSCS competence score ($f^2=0.42$), and 0.27 in the MSCS physical score ($f^2=0.51$), with large effect sizes for both. Cohen's f^2 values indicated a

small-to-medium effect size for all variables except MSCS academic scores, where a large effect size was observed.

ACCEPTED MANUSCRIPT

Discussion

To the best of our knowledge, this is the first study focusing on self-concept and its clinical correlates in patients with GTS at the transition age between adolescence and adulthood. Importantly, and differently from previous studies, we recruited patients within a narrow age range (15-19 years), with a mean age of 18 years, to ensure that our research findings referred to a patient population in the transitory period into adulthood. Interestingly, in our clinical sample, gender, socio-demographic variables, types of treatment interventions or tics/tic-related factors were not associated with decreased self-concept. However, we found a significantly lower report of self-concept in patients with the “GTS-plus” phenotype, i.e. patients diagnosed with at least one co-morbid psychiatric disorder. The presence of psychiatric co-morbidity appeared to affect to a greater extent the patients’ perception of their competence and academic skills, as well as their social and affective skills. This result is consistent with the findings of previous studies conducted on both children/adolescents and adults with GTS. Khalifa et al.⁽¹⁵⁾ found that physical and social aspects of self-perception were more strongly affected in young patients with GTS in the presence of co-morbid ADHD, whereas Thibert et al.⁽³⁰⁾ reported a low level of self-concept in adult patients with GTS who had co-morbid severe obsessive-compulsive symptoms.

With regard to the determinants of self-concept in our study sample, we found that the co-occurrence of anxiety symptoms exerts a negative impact on every self-concept domain assessed by the MSCS, and above all on the social and affective domains. Our results showed that self-concept could also be influenced by affective symptoms, with possible effects on the physical, affective, competence, and social domains. These findings were in line with the results of our multiple regression analysis, which showed that STAI-Y2 and BDI-II scores were the strongest predictors of MSCS scores. In contrast to state anxiety, trait anxiety (as measured by STAI-Y2 scores) can be defined as a relatively enduring disposition to feel stress, worry, and discomfort, and has continuity with personality. This is highly relevant, given the possibly increased risk of personality difficulties, as well as developmental disorders, in the GTS population⁽³¹⁻³³⁾. Interestingly, the findings of the recent study by Hesapçioğlu et al.⁽¹⁶⁾ on a clinical sample of children and adolescents with chronic tic disorders also suggested a pivotal role for anxiety and depression in determining self-esteem and HR-QOL. A study of youth with chronic tic disorder by Hanks et al.⁽¹⁸⁾ found medium-to-large-sized associations between patients’ self-concept and co-morbid psychiatric disorders (mainly severity of ADHD, OCD, and depressive symptoms), patients’ self-concepts partially mediated the relationship between tic severity and depressive symptom severity, and the interaction between tic impairment and reliance on avoidant coping strategies moderated patients’ self-concepts. Taken

together, the results of these studies highlight the importance of addressing the psychological well-being of patients with GTS as a way to improve their self-concept throughout childhood, adolescence and early adulthood^(34,35). Of note, the findings of clinical studies published in the last few years suggested the potential efficacy of broader interventions focused on the adverse psychosocial consequences of tics^(36,37).

Our study has limitations. Firstly, the relatively small sample size of some of the patient subgroups could have affected our findings. This was a consequence of the highly specialist study setting. Secondly, referral bias needs to be taken into account as participants were recruited from a tertiary referral centre, with focus on patients with more severe tics and may therefore not be representative of the community population of people with GTS. Specifically, the population studied was atypical as none had evidence of developmental delay or disorder and the rate of psychiatric co-morbidity was markedly lower than might have been expected from other studies⁽²⁾. Thirdly, the use of self-report questionnaires is subject to social acceptability bias, which might reduce the validity of the collected data. Moreover, as in other chronic illnesses, it may be difficult to avoid the possibility that the effects of a tic disorder or the side effects of anti-tic medications may lead to misleading answers in questions relating to psychiatric symptoms. Likewise, the possibility of a cross-over between questions measuring affective symptoms and those relating to self-concept cannot be ruled out completely, despite the choice of a self-concept measure which has been previously used in clinical populations with neuropsychiatric conditions. This issue might have been particularly relevant for the BDI-II items addressing self-dislike and self-criticalness, which are intrinsic to self-concept. A further limitation of this study is intrinsic to its retrospective approach. Finally, although the gender distribution in our sample reflected the male:female ratio observed in the wider GTS population, the small number of female patients enrolled in our study might have introduced a bias, in consideration of the higher prevalence of anxiety and affective disorders in girls and women.

In conclusion, our investigation of patient-reported self-concept in patients with GTS at the transition between adolescence and young adulthood highlighted the possible roles of anxiety and affective symptoms in influencing the psychological well-being of this patient population. Further research is needed to replicate these preliminary findings in larger samples both from specialist clinics and in the community. Self-concept and psychiatric health may have an important impact upon the process of adjustment to a chronic disorder that may endure into adulthood. Well adjusted young people entering adult life with GTS may enter a virtuous cycle in which their capacity to live with their tics both reduces their risk of psychiatric decompensation and cuts their need for ongoing treatment, with all its attendant risks. Since effective behavioural and pharmacological interventions are currently available for both anxiety and affective disorders, routine screening for

these co-morbid conditions should be recommended in all patients with GTS seen at transition clinics from paediatric to adult care. Finally, should our findings be confirmed by the results of future studies, novel treatment approaches might need to be implemented, as the efficacy of traditional interventions in trait anxiety may be less reliable.

Acknowledgments

Gratitude is expressed to the Tourette Association of America and Tourettes Action-UK for their continuing support.

Ethical standards

All human studies have been approved by the appropriate ethics committee and have therefore been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments. All participants gave their informed consent prior to their inclusion in the study.

Conflict of interest

The authors declare that they have no conflict of interest.

References

- [1] American Psychiatric Association (2013) Diagnostic and Statistical Manual for Mental Disorders, Fifth edition (DSM-5). American Psychiatric Publishing.
- [2] Cavanna AE, Seri S (2013) Tourette's syndrome. *Br Med J* 347:f4964.
- [3] Bloch MH, Peterson BS, Scahill L, Otko J, Katsoyich L, Zhang H, Leckman JF (2006) Adulthood outcome of tic and obsessive-compulsive symptom severity in children with Tourette syndrome. *Arch Pediatr Adolesc Med* 160:65-69.
- [4] Hassan N, Cavanna AE (2012) The prognosis of Tourette syndrome: Implications for clinical practice. *Funct Neurol* 27:23-27.
- [5] Robertson MM (2006). Tourette syndrome and affective disorders: An update. *J Psychosom Res* 61:349-358.
- [6] Frank MC, Piedad J, Rickards H, Cavanna AE (2011) The role of impulse control disorders in Tourette syndrome: An exploratory study. *J Neurol Sci* 310:276-278.
- [7] Chen K, Budman CL, Diego Herrera L, Witkin JE, Weiss NT, Lowe TL, Freimer NB, Reus VI, Mathews CA (2013) Prevalence and clinical correlates of explosive outbursts in Tourette syndrome. *Psychiatry Res* 205:269-275.
- [8] Cavanna AE, David K, Orth M, Robertson MM (2012) Predictors during childhood of future health-related quality of life in adults with Gilles de la Tourette syndrome. *Eur J Paediatr Neurol* 16:605-612.
- [9] Cavanna AE, David K, Bandera V, Termine C, Balottin U, Schrag A, Selai C (2013) Health-related quality of life in Gilles de la Tourette syndrome: A decade of research. *Behav Neurol* 27:83-93.
- [10] Packer LE (2005) Tic-related school problems: Impact on functioning, accommodations, and interventions. *Behav Modif* 29:876-899.

- [11] Abwender DA, Como PG, Kurlan R, Parry K, Fett KA, Cui L, Plumb S, Deeley C (1996) School problems in Tourette's syndrome. *Arch Neurol* 53:509-511.
- [12] Hoekstra PJ, Steenhuis MP, Troost PW, Korf J, Kallenberg CG, Minderaa RB (2004) Relative contribution of attention-deficit hyperactivity disorder, obsessive-compulsive disorder, and tic severity to social and behavioral problems in tic disorders. *J Dev Behav Pediatr* 25:272-279
- [13] Gorman DA, Thompson N, Plessen KJ, Robertson MM, Leckman JF, Peterson BF (2010) Psychosocial outcome and psychiatric comorbidity in older adolescents with Tourette syndrome: Controlled study. *Br J Psychiatry* 19:36-44.
- [14] Piedad JCP, Cavanna AE (2016) Depression in Tourette syndrome: A controlled and comparison study. *J Neurol Sci* 364:128-132.
- [15] Khalifa N, Dalan M, Rydell AM (2010) Tourette syndrome in the general child population: Cognitive functioning and self- perception. *Nord J Psychiatry* 64:11-18.
- [16] Hesapçioğlu ST, Tural MK, Kandil S (2014) Quality of life and self-esteem in children with chronic tic disorder. *Turk Pediatri Ars* 49:323-332.
- [17] Bracken B (1992) *Multidimensional Self- Concept Scale*. Austin, TX: PRO-ED.
- [18] Hanks CE, McGuire JF, Lewin AB, Storch EA, Murphy TK (2016) Clinical correlates and mediators of self-concept in youth with chronic tic disorders. *Child Psychiatry Hum Dev* 47:64-74.
- [19] Evans J, Seri S, Cavanna AE (2016) The effects of Gilles de la Tourette syndrome and other chronic tic disorders on quality of life across the lifespan: A systematic review. *Eur Child Adolesc Psychiatry* 25:939-948.
- [20] Leckman JF, Riddle MA, Hardin MT, Ort SI, Swartz KL, Stevenson J, Cohen DJ (1989) The Yale Global Tic Severity Scale: Initial testing of a clinician-rated scale of tic severity. *J Am Acad Child Adolesc Psychiatry* 28:566-573.

- [21] Scahill L, Riddle MA, McSwiggin-Hardin M, Ort SI, King RA, Goodman WK, Cicchetti D, Leckman JF (1997) Children's Yale-Brown Obsessive Compulsive Scale: Reliability and validity. *J Am Acad Child Adolesc Psychiatry* 36: 844-852.
- [22] Cavanna AE, Piedad JCP (2013) Clinical rating instruments in Tourette's syndrome. In: *Tourette's Syndrome* (eds Martino D, Leckman JF), pp. 411-438.
- [23] Woods DW, Piacentini J, Chang S, Deckersbach T, Ginsburg G, Peterson A, Scahill LD, Walkup JT, Wilhelm S (2008) *Managing Tourette Syndrome: A Behavioral Intervention for Children and Adults (Therapist Guide)*. New York, NY: Oxford University Press.
- [24] Woods DW, Piacentini J, Himle MB, Chang S (2005) Premonitory Urge for Tics Scale (PUTS): Initial psychometric results and examination of the premonitory urge phenomenon in youths with tic disorders. *J Dev Behav Pediatr* 26:397-403.
- [25] Esposito M, Gallai B, Parisi L, Castaldo L, Marotta R, Lavano SM, Mazzotta G, Roccella M, Carotenuto M (2013) Self-concept evaluation and migraine without aura in childhood. *Neuropsychiatr Dis Treat* 9:1061-1066.
- [26] Gagliano A, Lamberti M, Siracusano R, Ciuffo M, Boncoddio M, Maggio R, Rosina S, Cedro C, Germanò E (2014) A comparison between children with ADHD and children with epilepsy in self-esteem and parental stress level. *Clin Pract Epidemiol Ment Health* 10:176-183.
- [27] March JS, Parker JD, Sullivan K, Stallings P, Conners CK (1997) The Multidimensional Anxiety Scale for Children (MASC): Factor structure, reliability, and validity. *J Am Acad Child Adolesc Psychiatry* 36:554-565.
- [28] Spielberger CD, Gorsuch RL, Lushene R, Vagg PR, Jacobs GA (1983) *Manual for the State-Trait Anxiety Inventory*. Palo Alto, CA: Consulting Psychologists Press.
- [29] Beck AT, Steer RA, Brown GK (1996) *Beck Depression Inventory-Second Edition Manual*. San Antonio, TX: The Psychological Corporation.

- [30] Thibert AL, Day HI, Sandor P (1995) Self-concept and self-consciousness in adults with Tourette syndrome. *Can J Psychiatry* 40:35-39.
- [31] Eddy CM, Rickards HE, Critchley HD, Cavanna AE (2013) A controlled study of personality and affect in Tourette syndrome. *Compr Psychiatry* 54:105-110.
- [32] Trillini MO, Müller-Vahl KR (2015) Patients with Gilles de la Tourette syndrome have widespread personality differences. *Psychiatry Res* 228:765-773.
- [33] Balottin L, Selvini C, Luoni C, Mannarini S, Chiappedi M, Seri S, Termine C, Cavanna AE (2016). Personality profile of male adolescents with Tourette syndrome: A controlled study. *J Child Neurol* 31:294-299.
- [34] Lewin AB, Storch EA, Conelea CA, Woods DW, Zinner SH, Budman CL (2011) The roles of anxiety and depression in connecting tic severity and functional impairment. *J Anx Disord* 25:164-168.
- [35] Trillini MO, Müller-Vahl KR (2015) Narcissistic vulnerability is a common cause for depression in patients with Gilles de la Tourette syndrome. *Psychiatry Res* 230:695-703.
- [36] McGuire JF, Arnold E, Park JM, Nadeau JM, Lewin AB, Murphy TK, Storch EA (2015) Living with tics: Reduced impairment and improved quality of life for youth with chronic tic disorders. *Psychiatry Res* 225: 571-579.
- [37] Leclerc JB, O'Connor KP, J-Nolin G, Valois P, Lavoie ME (2016) The effect of a new therapy for children with tics targeting underlying cognitive, behavioral, and physiological processes. *Front Psychiatry* 7:135.

TABLES

ACCEPTED MANUSCRIPT

Table 1. Self-concept ratings (mean values \pm SD) in patients with Gilles de la Tourette syndrome grouped by clinical variables.

Table 2. Results of multiple linear regression analysis of clinical determinants of self-concept in patients with Gilles de la Tourette syndrome.

ACCEPTED MANUSCRIPT

Table 1. Self-concept ratings (mean values \pm SD) in patients with Gilles de la Tourette syndrome grouped by clinical variables.

		MSCS social	MSCS competence	MSCS affect	MSCS academic	MSCS family	MSCS physical	MSCS total
BDI-II	p	0.0428	0.0322	0.0157	0.1103	1.0000	0.0090	0.0671
Neg	n=15	104.33 \pm 18.36	101.87 \pm 18.22	99.53 \pm 13.79	93.67 \pm 22.00	103.20 \pm 12.29	101.00 \pm 11.23	100.47 \pm 16.62
Pos	n=6	84.00 \pm 19.87	81.83 \pm 13.11	80.50 \pm 9.73	78.83 \pm 10.48	101.83 \pm 7.83	82.83 \pm 8.57	82.83 \pm 12.17
MASC	p	0.0020	0.0894	0.0226	0.4175	0.0965	0.0107	0.0086
Neg	n=12	108.83 \pm 14.47	102.50 \pm 19.74	99.92 \pm 14.53	92.42 \pm 23.93	105.50 \pm 8.90	102.00 \pm 11.20	102.42 \pm 15.13
Pos	n=8	80.63 \pm 16.69	85.75 \pm 14.84	83.75 \pm 11.57	83.38 \pm 14.20	96.63 \pm 11.15	85.88 \pm 11.27	83.25 \pm 14.46
CO-MORB	p	0.0224	0.0073	0.0124	0.0082	0.0593	0.2043	0.0137
Neg	n=8	111.88 \pm 16.91	109.75 \pm 16.08	105.38 \pm 13.20	105.38 \pm 17.71	109.75 \pm 11.65	101.13 \pm 10.70	108.00 \pm 15.48
Pos	n=13	90.31 \pm 18.63	87.77 \pm 15.87	87.15 \pm 12.34	79.62 \pm 15.24	98.54 \pm 8.45	92.54 \pm 14.08	87.69 \pm 13.60
PUTS	p	0.4254	0.0886	0.2177	0.0758	0.7168	0.1277	0.1575
Neg	n=13	101.31 \pm 22.44	101.85 \pm 20.01	97.77 \pm 16.48	96.54 \pm 18.59	102.69 \pm 12.73	99.85 \pm 11.56	99.92 \pm 18.22
Pos	n=8	94.00 \pm 17.56	86.88 \pm 13.67	88.13 \pm 11.78	77.88 \pm 18.50	103.00 \pm 8.32	89.25 \pm 14.07	88.13 \pm 13.44
STAI-Y1	p	0.0015	0.0049	0.0062	0.0432	0.0192	0.0807	0.0026
Neg	n=13	109.30 \pm 15.79	105.77 \pm 16.84	101.54 \pm 14.05	97.46 \pm 18.83	107.15 \pm 11.36	100.69 \pm 11.97	104.38 \pm 14.87
Pos	n=7	78.00 \pm 12.94	80.43 \pm 10.47	80.71 \pm 7.52	75.14 \pm 16.96	95.43 \pm 6.19	87.71 \pm 12.98	79.43 \pm 8.48
STAI-Y2	p	0.0073	0.0015	0.0093	0.0208	0.0430	0.0207	0.0017
Neg	n=14	106.86 \pm 17.75	105.36 \pm 16.11	100.36 \pm 14.46	97.36 \pm 17.71	106.29 \pm 10.52	101.26 \pm 10.19	103.57 \pm 14.46
Pos	n=6	78.50 \pm 14.01	77.17 \pm 7.36	80.00 \pm 6.66	71.67 \pm 16.72	95.50 \pm 9.77	84.17 \pm 13.53	77.17 \pm 7.36

Statistically significant values ($p \leq 0.05$; Mann-Whitney U test) shown in bold.

Abbreviations. MSCS, Multidimensional Self Concept Scale; BDI-II, Beck Depression Inventory-II; MASC, Multidimensional Anxiety Scale for Children; CO-MORB, presence of at least one co-morbid psychiatric disorder ("GTS-plus"); PUTS, Premonitory Urge for Tics Scale; STAI-Y1, State-Trait Anxiety Inventory-State Anxiety; STAI-Y2, State-Trait Anxiety Inventory-Trait Anxiety.

ACCEPTED MANUSCRIPT

Table 2. Results of multiple linear regression analysis of clinical determinants of self-concept in patients with Gilles de la Tourette syndrome.

	OCD		ADHD		BDI-II		STAI-Y2		MASC		Adj R ²	AIC
	coeff	95% CI Cohen's f ²	coeff	95% CI Cohen's f ²	coeff	95% CI Cohen's f ²	coeff	95% CI Cohen's f ²	coeff	95% CI Cohen's f ²		
MSCS social	-0.99	-15.7 - 13.7 0.002	-9.16	-24.2 - 5.8 0.134	0.06	-0.28 - 0.41 0.012	-0.37	-0.68 - -0.05 0.598	-0.41	-1.13 - 0.31 0.181	0.66	154.4
MSCS competence	-2.89	-12.3 - 6.5 0.033	-8.09	-17.7 - 1.51 0.254	-0.24	-0.46 - -0.02 0.409	-0.27	-0.48 - -0.07 0.618	-0.03	-0.49 - 0.44 0.024	0.84	137.4
MSCS affect	-1.49	-11.4 - 8.4 0.008	-2.47	-12.6 - 7.6 0.021	-0.21	-0.44 - 0.02 0.291	-0.22	-0.43 - -0.01 0.449	0.02	-0.47 - 0.50 0.147	0.72	139.3
MSCS academic	-14.95	-29.1 - -0.84 0.403	-11.61	-26.0 - 2.76 0.234	-0.27	-0.60 - 0.07 0.232	-0.19	-0.49 - 0.11 0.353	0.20	-0.50 - 0.89 0.023	0.69	152.7
MSCS family	-5.25	-15.7 - 5.2 0.090	-5.28	-15.9 - 5.4 0.088	0.07	-0.18 - 0.31 0.025	-0.23	-0.45 - -0.01 0.389	0.19	-0.32 - 0.70 0.039	0.34	141.4
MSCS physical	1.87	-8.7 - 12.4 0.011	6.79	-3.91 - 17.5 0.145	-0.27	-0.52 - -0.02 0.433	-0.07	-0.29 - 0.15 0.046	-0.18	-0.70 - 0.33 0.142	0.59	141.6
MSCS total	-5.09	-14.0 - 3.79 0.118	-5.25	-14.3 - 3.80 0.121	-0.14	-0.35 - 0.07 0.163	-0.29	-0.48 - -0.10 0.987	-0.02	-0.45 - 0.42 0.125	0.82	135.2

Maximum Variance Inflation Factor for the regression model = 3.67; statistically significant values ($p \leq 0.05$; multiple linear regression analysis) shown in bold.

Abbreviations. OCD, obsessive-compulsive disorder; ADHD, attention-deficit and hyperactivity disorder; BDI-II, Beck Depression Inventory-II; STAI-Y2, State-Trait Anxiety Inventory-Trait Anxiety; MASC, Multidimensional Anxiety Scale for Children; MSCS, Multidimensional Self Concept Scale; Adj R² = adjusted R²; AIC = Akaike Information Criterion; coeff = coefficient of the multiple linear regression model; 95% CI = 95% Confidence Interval; Cohen's f² = effect size for multiple regression.

ACCEPTED MANUSCRIPT

Gilles de la Tourette syndrome (GTS) has an impact on quality of life and self-esteem

This was the first study to investigate self-concept in GTS in late adolescence

Lower self-concept was associated with the presence of comorbid psychiatric disorders

Trait anxiety symptoms were the main determinants of self-concept in our sample